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Hydrocolpos: Etiology, Diagnosis and Management

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1. Abstract

1.1. Background

Hydrcolpos is a rare condition and commonly seen in newborns as a result of vaginal obstruction together with accumulation of secretion from the secretory glands of the uterine cervix and vagina leading to distension and dilatation of the vagina. The accumulated secretions can extend to the uterus leading to its dilation, a condition known as hydrometrical. There are several causes for hydrocolpos and early diagnosis and treatment are important ant to avoid associated complications.

1.2. Patients and Methods

We treated eight patients with hydrocolpos. Their medical records were reviewed for age at presentation, birth weight, clinical features, diagnosis and management.

1.3. Results

Eight female patients were treated. Seven of them presented with hydrocolpos immediately after birth and one presented at the age of 13.5 years after menarche and was diagnosed to have hematocolpos secondary to imperforated hymen. The remaining seven were diagnosed to have imperforated hymen in two, vaginal atresia in four and persistent cloaca in one. Abdominal ultrasound, CT scan and MRI when necessary for valuable investigations to confirm the diagnosis and plan treatment. All of them had obstructive uropathy on ultrasound except one. Two of them were found to have pyocolpos as a result of secondary infection of the hydrocolpos. All were operated on as newborns except one who presented at the age of 13.5 years and all made good recovery except one who had cloaca. This patient died two weeks postoperatively as a result of sepsis.

1.4. Conclusions

Congenital hydrocolpos is a rare condition. Hydrocolpos should be included in the differential diagnosis of any newborn presenting with a pelvic cystic mass. There are several causes for hydrocolpos and early diagnosis and proper management is important to avoid associated complications.

2. Introduction

Congenital hydrocolpos is a rare condition characterized by cystic dilatation of the vagina as a result of vaginal obstruction and accumulation of secretions from the secretary glands of the female cervix and vagina [1,2,3]. The vaginal distension with secretion accumulation is secondary to increased secretion by cervical mucous glands as a result of maternal hormone stimulation and in the presence of vaginal obstruction these secretions gradually accumulate in the vagina leading to its dilation [3,4,5]. Neonatal hydrocolpos develops as a result of several conditions including imperforated hymen, cloacal malformations and vaginal atresia [6,7,8]. Prenatal diagnosis and early newborn evaluation and imaging studies are important for early diagnosis and treatment of these patients. We report eight cases of hydrocolpos outlining aspects of diagnosis and treatment.

3. Case Reports

Case No. 1: A newborn female, a product of consanguineous marriage was referred to our hospital. She was a product of caesarian section with a birth weight of 3.6 kg. She presented with abdominal distension. She was diagnosed to have congenital heart disease and her antenatal ultrasound showed a pel-

vic mass. Clinically, she was found to have a lower abdominal mass arising from the pelvis and extending upwards (Figure 1). She was also found to have polydactyl and syndactyl involving her right hand and right foot (Figure 2a and 2b). She was also found to have an anterior ectopic anus and her echocardiography showed common atrio-ventricular canal with single atrium, moderate patent ductus arteriosum and severe atrio-ventricular regurgitation (Figure 3). Her abdominal ultrasound and CT-scan showed hydrometrocolpos with bilateral hydroureteronephrosis and left perinephric fluid collection. She was diagnosed to have Mckusick-Kaufman syndrome. She was operated on through a Pfannenstiel incision and found to have hydrometrocolpos secondary to low vaginal atresia. An abdomino-perineal vaginal pull through was done and anew vaginal opening was fashioned. Postoperatively, she did well and was discharged home to be followed up in the clinic.

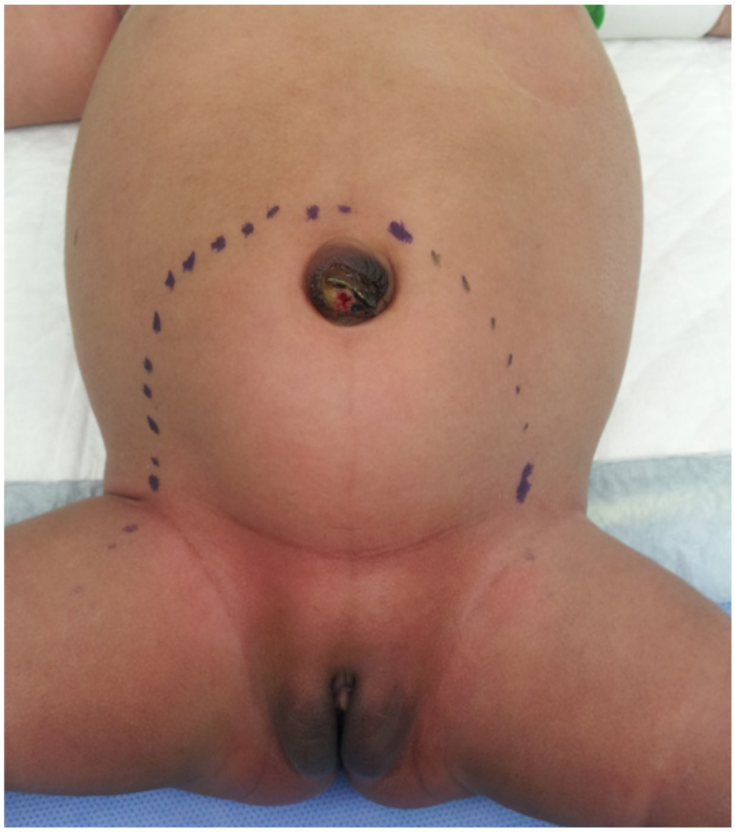
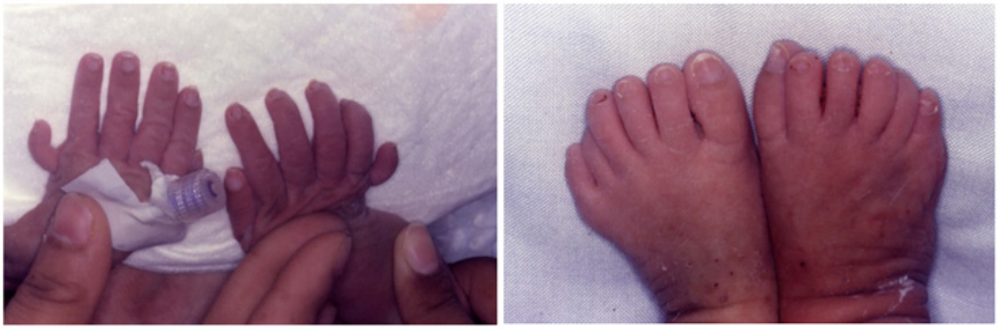


Figure 1: A clinical photograph showing a female newborn with abdominal distension and an abdominal mass arising from the pelvis and extending upwards into the peritoneal cavity



Figures 2a and 2b: Clinical photographs showing polydactly involving both hands and feet.

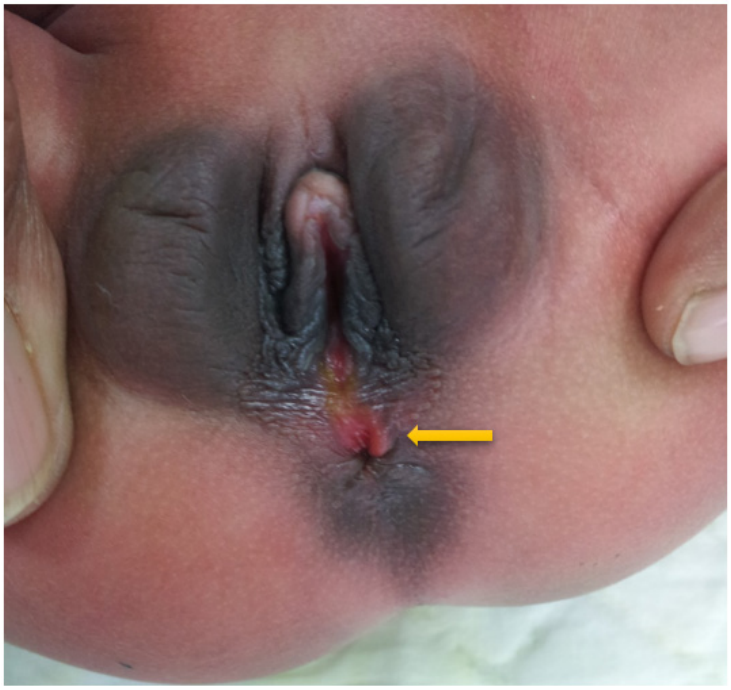


Figure 3: A clinical photograph of a newborn female with hydrocolpos and an associated anterior ectopic anus.

Case No. 2: a 4-day old female was referred to our hospital because of abdominal mass and a swelling protruding from her vaginal opening. Her birth weight was 3.2 kg and she was a product of full term normal vaginal delivery. Clinically, she was found to have anormal anal opening but there was a membrane covering the introitus and bulges during straining (Figure 4). She was also found to have a lower abdominal cystic mass arising from the pelvis and extending upwards into the peritoneal cavity. Abdominal and pelvic ultrasound and CT-scan confirmed the diagnosis of hydrocolpos. There was also hydroureter and hydronephrosis on the left side. The right kidney was normal. The diagnosis of imperforated hymen and hydrocolpos was made. She was operated on and the hymen was incised through a cruciate incision. The edges were trimmed and sutured with 5.0 vicryl sutures. About 250 of straw-colored fluid was drained. Postoperatively, she did well and was discharged home in good general condition. A follow up ultrasound showed resolution of the hydrocolpos and also the hydronephrotic changes on the left side.

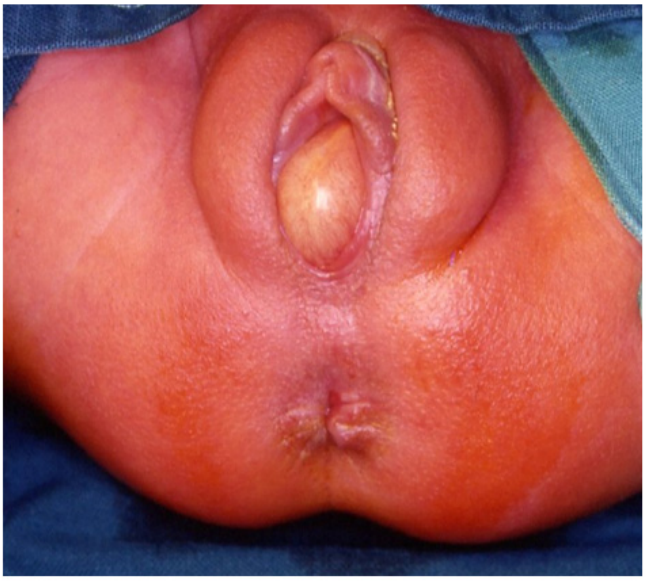
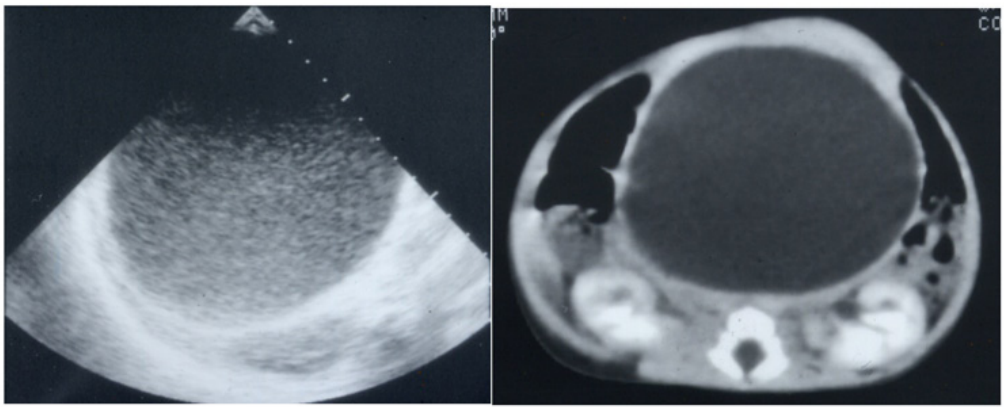


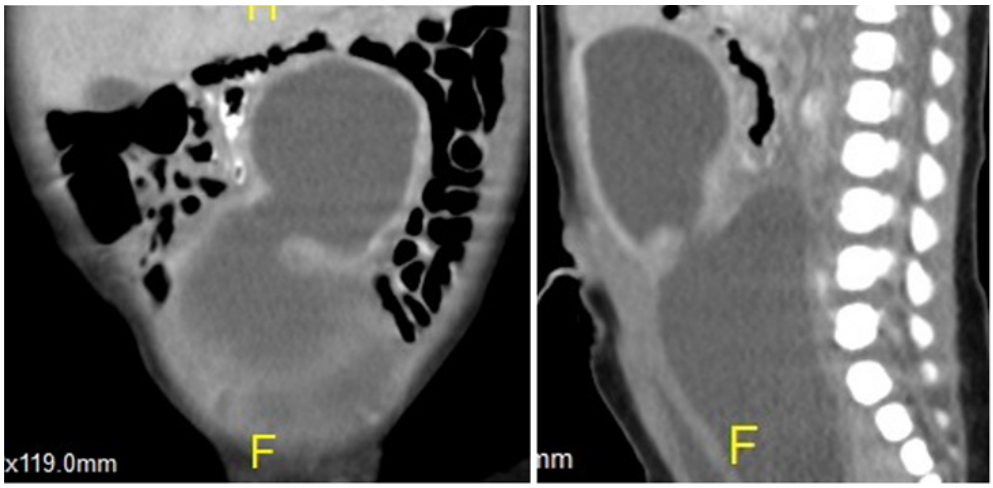
Figure 4: A clinical photograph of a newborn female with hydrocolpos. Note the imperforated hymen which is bulging from the introitus.

Case No. 3: A female newborn was referred to our hospital with an abdominal mass. She was a product of full term normal vaginal delivery. Her birth weight was 3 kg. Clinically she found to have a normal anal opening but there were no separate vaginal and urethral openings. Abdominal examination revealed a large cystic mass arising from the pelvis and extending upwards to above the umbilicus. Abdominal ultrasound, CT-scan and MRI showed a large cystic pelvi-abdominal mass (Figures 5a, 5b, 6a, 6b, 7a, 7b, 8a and 8b). The mass measured 11.5 X 8 X 6.5 cm. Both kidneys showed hydronephrosis. She was diagnosed to

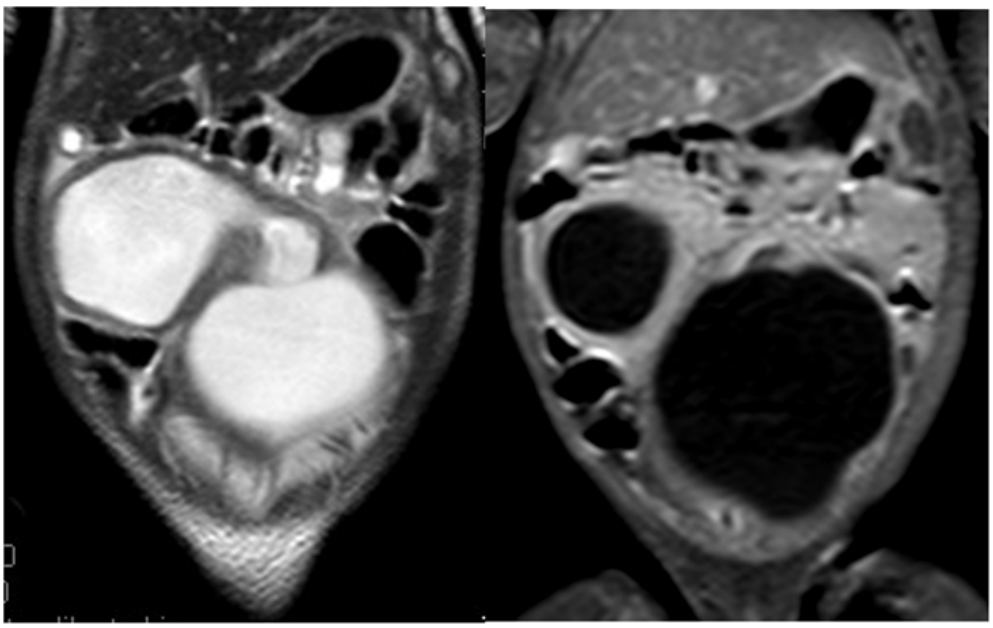
have hydrometrocolpos secondary to vaginal atresia. Examination under general anesthesia revealed no vaginal opening and no separated urethral opening but a common channel with the urethral opening into it. Exploration through a lower abdominal transverse incision showed a markedly distended vagina. An abdomino-vaginal pull through and a vaginoplasty was performed (Figures 9a and 9b). Postoperatively, she did well and was discharged home in a good general condition.



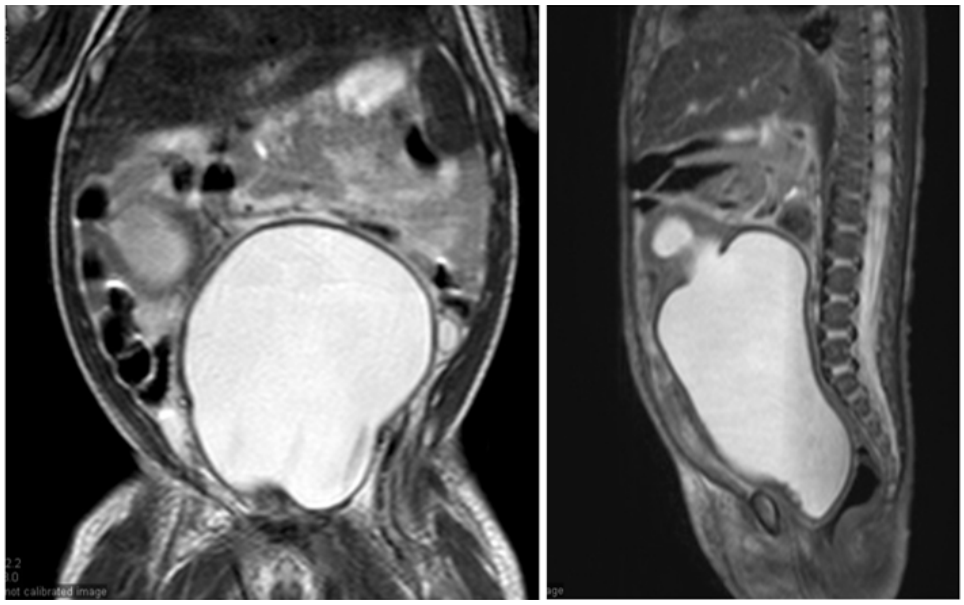
Figures 5a and 5b: Abdominal and pelvic ultrasound and CT scan showing a large cystic pelvic mass extending into the peritoneal cavity. This represents the markedly dilated vagina.



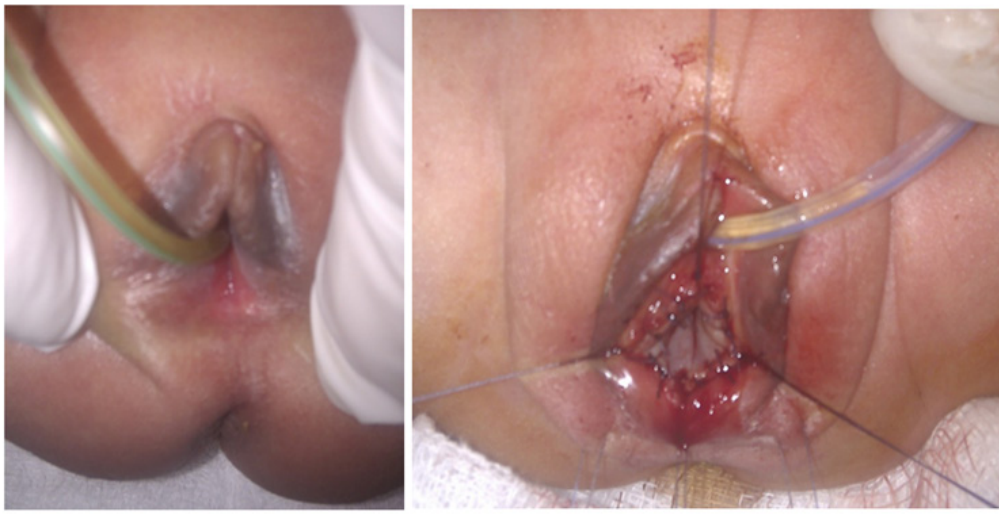
Figures 6a and 6b: Abdominal and pelvic MRI scan showing a large cystic pelvic mass extending into the peritoneal cavity. This represents the markedly dilated vagina. Note also the dilated uterus forming a hydrometrocolpos.



Figures 7a and 7b: Abdominal and pelvic MRI showing a large cystic pelvic mass extending into the peritoneal cavity. This represents the markedly dilated vagina. Note also the dilated uterus as a result of the accumulated fluid extending from the vagina and extending to distend the uterus forming a hydrometrocolpos.



Figures 8a and 8b: Abdominal and pelvic MRI showing a large cystic pelvic mass extending into the peritoneal cavity. This represents the markedly dilated vagina.



Figures 9a and 9b: Clinical and intraoperative photographs showing no vaginal opening and a newly constructed vaginal opening following an abdomino-perineal vaginal pull-through.

Case No. 4: A 13.5-year-old female was admitted to our hospital because of repeated attacks of lower abdominal pain and backache for the last 5 months. She also gave a history of urine retention, frequency and dysuria. Clinically, she was found to have imperforated hymen which was bulging. Abdominal and pelvic ultrasound showed a normal uterus, normal ovaries but a markedly distended vagina with fluid. The diagnosis of imperforated hymen with hematocolpos was made. She was operated on and upon incision of the hymen about 200 ml of altered blood was drained. Postoperatively, she did well and on follow up 3 months postoperatively she was well with normal looking perineum.

Case No. 5: A newborn female was referred to our hospital because of imperforated anus and an abdominal mass. She was after 36 weeks gestation and her birth weight was 1.65 kg. Clinically, she was found to have imperforated anus and a large abdominal mass arising from the pelvis and extending to above the umbilicus (Figure 10). There was only one common opening in the perineum and she was diagnosed to have a persistent cloaca. Abdominal ultrasound showed a markedly distended uterus and vagina and bilateral hydroureteronephrosis. She was operated on through a lower transverse abdominal incision. The vagina was markedly distended. There was a single uterus which was slightly distended and two Fallopian tubes. The distended vagina was opened and about 250 ml of fluid was drained. A vaginostomy was made and a sigmoid colostomy was fashioned. The urinary bladder was also drained. Postoperatively, the patient did well for 5 days but then she became septic and died two weeks postoperatively.

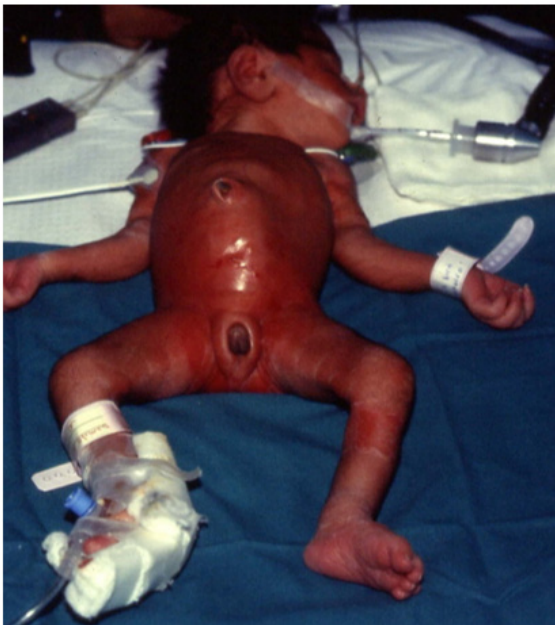
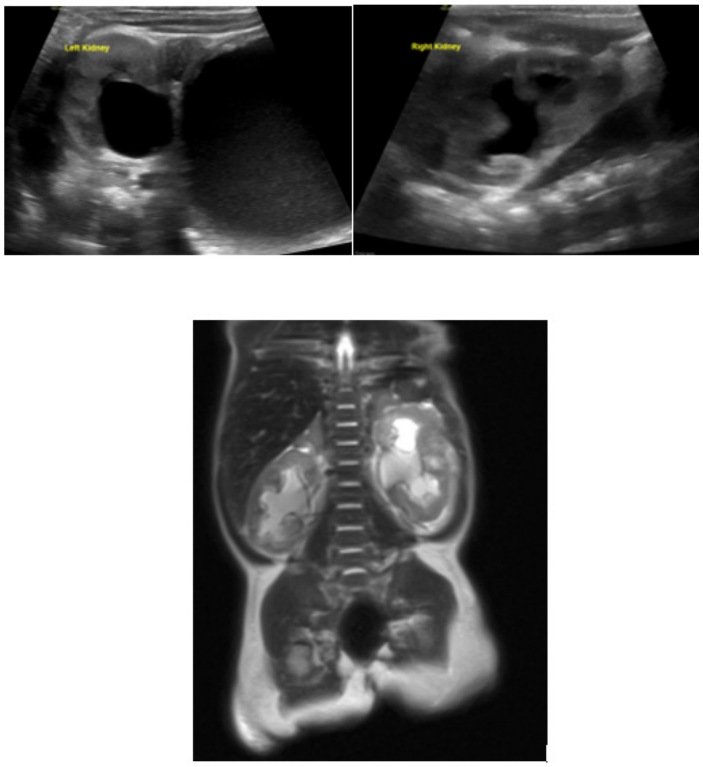


Figure 10: A clinical photograph of a female newborn who presented with one perineal opening and abdominal distension. She was diagnosed to have persistent cloaca and hydrocolpos.

Case No. 6: A 5 days old female was referred to out hospital because of a bulge in the perineum at the site of the vaginal opening. Her birth weight was 3.2 kg Clinically, she was found to have a lower abdominal cystic mass which was tender and arising from the pelvis. Her external genitalia showed an imperforated hymen which was budging. Abdominal and pelvic ultrasound showed a cystic swelling arising from the pelvis and consistent with a hydrocolpos. She was operated on and the imperforated hymen was opened via a small cruciate incision. The edges were trimmed and a large amount of creamy fluid came out. A swab was taken for culture and sensitivity which grew Klebsiella and Enterococci confirming the diagnosis of pyocolpos secondary to imperforated hymen. She was treated with antibiotics and did well and was discharged home in a good general condition.

Case No. 7: A one-week-old female was referred to our hospital because of abdominal distension. She was a product of full term normal vaginal delivery. Her birth weight was 3 kg. Examination revealed a sick looking baby with a large, tender cystic swelling arising from the pelvis and extending to just above the umbilicus. There was anormal anal opening but no vaginal opening and only a common vaginal and urethral opening where she was passing urine from. No other abnormalities detected. Abdominal ultrasound revealed a large cystic mass of mixed fluid content arising from the pelvis and extending upwards. This was confirmed by CT-scan and both kidneys showed hydronephrotic changes. Under general anesthesia, the common channel was catharized and this drained urine. Exploration through a lower transverse abdominal incision. The vagina was opened and about 450 ml of yellowish fluid was drained. Culture of this fluid grew E. coli and enterococcus faecalis. She was also found to have distal vaginal atresia. A catheter was passed and guided from the common channel into the vagina to drain the pyocolpos and the patient was treated with intravenous antibiotics. Postoperatively, she did well and six weeks later she underwent and abdomino-vaginal pull-through and a new vaginoplasty was performed. Postoperatively she did well and was discharged home in a good general condition.

Case No. 8: A term female neonate was born at 37 weeks' gestation and a birth weight of 3.09 kg was admitted to our hospital. She was diagnosed to hydrometrocolpos associated with obstructive uropathy by antenatal ultrasound. Clinically, there was no vaginal opening and was found to have markedly distended abdomen with a palpable large cystic abdominal mass arising from the pelvis and extending into the abdominal cavity. She was also found to have postaxial polydactyly involving all four limbs. Postdelivery her renal function and electrolytes were normal and abdominal and pelvic ultrasound and MRI showed Severe bilateral hydronephrosis, markedly dilated vagina, urinary bladder and uterus (Figure 11a, 11b and 11c). She was diagnosed to have McKusick–Kaufman syndrome. Her initial echocardiogram was normal. Her abdominal MRI showed a markedly dilated vagina and uterus (Figure 12). She underwent diagnostic cystoscopy with Foley catheter insertion and percutaneous vaginal pigtail catheter drainage. This improved her urine output and also the hydroureter and hydronephrosis as there was significant reduction in the size of the vagina and uterus. Once her condition was stabilized, she was operated on and an abdomino-perineal vaginal pull-through was done. Postoperatively she did well and was discharged home on the seventh postoperative day.



Figures 11a, 11b and 11c: Abdominal and pelvic ultrasound and MRI showing bilateral hydroureter and hydronephrosis in a female patient with hydrocolpos.

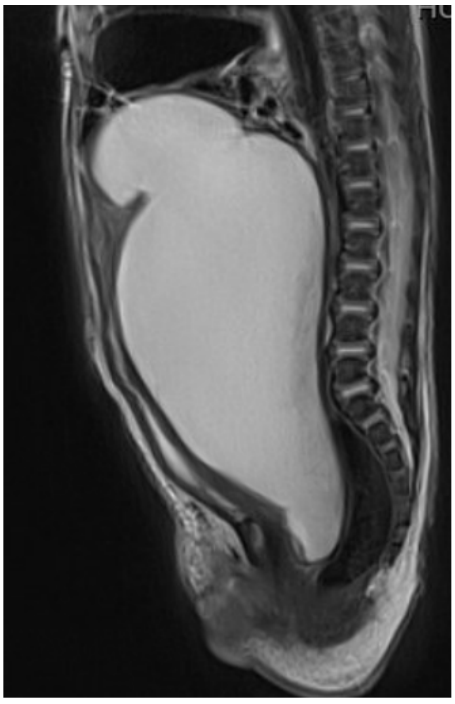


Figure 12: Abdominal and pelvic MRI showing a large cystic pelvic mass extending into the peritoneal cavity. This represents the markedly dilated vagina.

4. Discussion

Hydrocolpos is a rare condition with an estimated incidence of 1/16,000–30,000 female births [1,2]. Hydrocolpos is an accumulation of fluid in the vagina and if this extend to involve the uterus it is called hydrometrocolpos. Hydrocolpos can be diagnosed antenatally as a result of the wide use of antenatal ultrasound [8,9,10]. Hydrocolpos should be suspected antenatally when a cystic mass is seen in a female arising from the pelvis and extending upwards into the abdominal cavity.

Hydrocolpos is divided into two types depending on the accumulated fluid in the vagina [3]. The secretory type is the most common type and it is due to vaginal obstruction with accumulation of mucoid material secreted by the glands of the cervix and vagina as a result of in utero maternal hormonal stimulation. The common causes of vaginal obstruction include imperforate hymen and vaginal atresia or vaginal septum [4,5,11]. The urinary type is much rarer and this is seen in newborns with cloaca malformation [6,7]. This is a very severe and rare malformation

which is characterized by a single common perineal opening where the rectum, the vagina, and the urinary tract open. This single partially obstructed common urogenital canal leads to reflux of urine from the urinary bladder into the vagina leading to its distension. In these cases, it is also possible for the secretions from the cervix glands to accumulate in the partially obstructed vagina leading to its distension. One of the serious complications of hydrocolpos is the development of pyocolpos as a result of secondary infection of the hydrocolpos [4,12]. Pyocolpos can lead to sepsis of newborns which is known to be associated with a high mortality. We advocate early definitive surgery for these patients to avoid subsequent complications and also relive the pressure on the urinary tract caused by the markedly distended vagina. Patients who are not fit to undergo early definitive surgery should have a temporary drainage of the hydrocolpos to relive the distension and the pressure on the urinary system. Temporary drainage of the hydrocolpos can be done by placement of a catheter under ultrasound guidance by an interventional radiologist.

Hydrometrocolpos is a rare condition and now commonly diagnosed prenatally at ultrasound screening. The distended vagina may attain a large size presenting postnatally as a pelvic cystic mass, if large enough, may cause compression on both the gastrointestinal tract and the urinary system. The cystic mass arises in the pelvis and extend upwards into the peritoneal cavity pushing the intestines upwards and this causes splinting of the diaphragm causing respiratory distress. As a result of compression on the urinary system these patient present with partial obstruction causing hydroureters and hydronephrosis. Early diagnosis and treatment improve the prognosis especially in those with associated obstructive uropathy [13]. Once the accumulated fluid is drained, the pressure on the urinary system will be alleviated and this will lead to improvement in the hydroureter and hydronephrosis. Any vaginal obstruction like imperforate hymen, complete transverse vaginal septum, or partial vaginal agenesis may cause hydrocolpos and as the fluid accumulate in the vagina, it then extends into the uterus causing hydrometrocolpos. The wall of the uterus is thick and not distensible as the vagina. Once the fluid accumulates also in the uterus, it can pass into the peritoneal cavity through the fallopian tubes causing ascites. In the absence of significant amounts of accumulated secretions, these cases can be missed in the neonatal period and are diagnosed later during childhood, early adolescence, or after the menarche when they present with lower abdominal pain and hematocolpos as a result of accumulation of menstruation blood in the vagina as was the case in one of our patients. The cause of the large amount of accumulated fluid in some of these patients and not the others is not known. It was postulated that this is due to either remarkable concentrations of maternal estrogens in the mucosal lining of the fetal genital tract or very sensitive estrogen receptors in some of these patients. The effect of maternal estrogen can continue in some of these patients postnatally as was seen in our patients who had drainage of the hydrocolpos but the fluid reaccumulated once the catheter was removed. The duration of the maternal hormonal effect is not exactly known but once a definitive surgery was done there was no more secretions draining through the new vaginoplasty or following hymenotomy in those with imperforated hymen.

Vaginal atresia can be associated with Mckusick-Kaufman syndrome. McKusick-Kaufman syndrome is characterized by the triad of hydrometrocolpos, postaxial polydactyly and congenital heart disease [11,14,15]. This condition was first described in the Amish population [15]. McKusick-Although Mckusick-Kaufman syndrome is characterized by the triad of hydrometrocolpos, postaxial polydactyly and congenital heart disease, it was found that hydrometrocolpos is present in 80–95% of cases; postaxial polydactyly is present in 90% of patients; and congenital heart diseases have been described in 15%-20% of patients. Two of our patients had Mckusick-Kaufman syndrome, one of them had the classic triad of the syndrome while the second one had hydrocolpos and postaxial polydactyly but no evidence of congenital heart disease. Clinically, the patient may present with a large, cystic abdominal mass which can be sufficiently large to cause intestinal obstruction, urinary outflow obstruction leading to hydroureter and hydronephrosis, and/or elevation of the diaphragm resulting in breathing difficulties, associated with postaxial polydactyly and congenital heart defects. Both our patients had hydroureter and hydronephrosis. Our patient with congenital heart disease had common atrio-ventricular (A-V) canal with single atrium, moderate patent ductus arteriosus and severe A-V regurgitation. Other cardiac malformations include ventricular septal defect, atrial septal defect, small aorta and hypoplastic left ventricle, tetralogy of Fallot, and patent ductus arteriosus have been described in patients with Mckusick-Kaufman syndrome. The severity of the associated congenital heart malformations determines the final outcome. Cardiac evaluation and echocardiogram should form part of the preoperative investigations of these patients to accurately define the associated cardiovascular malformations. Additional less commonly associated abnormalities include imperforate anus, rectovaginal or vesicovaginal fistulae, Hirschsprung's disease, anterior ectopic anus and malrotation [11,15]. One of our patients had also an associated anterior ectopic anus. It is important to define anatomic abnormality leading to the hydrometrocolpos. We found CT- scan valuable in this regard. MRI has been reported to be more valuable than ultrasound and CT-scan in delineating the vaginal anatomical defect and the hydroureter and hydronephrosis [10,16]. Genitography is an unnecessary invasive investigation that may be harmful leading to secondary infection with subsequent pyometrocolpos. Pyometrocolpos

which is a serious complication calls for early and rapid evaluation and treatment of these patients.

There are various causes of hydrcolpos, and according to the etiology, surgical management can be simple or more complicated [17]. Hydrocolpos can be associated with genitourinary anomalies from persistent urogenital sinus to cloacal malformation [6,7]. The treatment of an imperforated hymen is simple, curative and should not be delayed. The timing of the surgical procedures for the other causes of hydrcolpos is still controversial. There are those who advocate postponing the definite procedure. We advocate early surgical correction to prevent complications of hydrocolpos and avoid secondary infection with the development of pyocolpos [4,12]. Two of our patients presented to our hospital with pyocolps. In one of them this was managed with drainage and antibiotics followed by definitive surgery 6 weeks later. The other patient had imperforated hymen and was treated with hymenoplasty which provided adequate drainage of the pyocolpos. The surgical drainage of hydrocolpos dramatically improve the hydroureter and hydronephrosis. Patients with vaginal atresia are best treated with abdomino-perineal vaginal pull through procedure. This is a definitive and curative procedure and one advantage of doing this early is the markedly distended and dilated vagina which makes the vaginal pull-through much easier. Patients who are not fit to undergo early vaginal pull-through can be managed temporarily by draining the hydrocolpos with an indwelling transabdominal vaginostomy tube for continuous drainage until the definitive repair. This can be done by interventional radiologist under ultrasound guidance. The management of patients with persistent cloaca is complex and should be done in specialized centers. These patients can be managed initially with a drainage vesicostomy and a colostomy to drain the associated hydrocolpos and also allow them to feed and pass stools. The definitive procedure can be delayed and should be done in centers with enough experience in managing these patients. These patients are at increased risk of sepsis and death as happened in our patient.

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